ELSEVIER

Contents lists available at ScienceDirect

Journal of Pediatric Surgery

journal homepage: www.elsevier.com/locate/jpedsurg



Outcomes after enterostomies in children with and without motility disorders: A description and comparison of postoperative complications



Mana H. Vriesman ^{a,*}, Najia Noor ^a, Ilan J. Koppen ^a, Carlo Di Lorenzo ^b, Justin R. de Jong ^c, Marc A. Benninga ^a

- a Department of Pediatric Gastroenterology and Nutrition, Emma Children's Hospital, Amsterdam UMC, University of Amsterdam, Amsterdam, the Netherlands
- ^b Division of Gastroenterology, Hepatology and Nutrition, Nationwide Children's Hospital, Columbus, Ohio, USA
- ^c Department of Pediatric Surgery, Emma Children's Hospital, Amsterdam UMC, University of Amsterdam, Amsterdam, the Netherlands

ARTICLE INFO

Article history: Received 30 October 2019 Received in revised form 28 April 2020 Accepted 15 May 2020

Key words: Stoma Enterostomy Ileostomy Colostomy Pediatric Motility disorders

ABSTRACT

Purpose: To provide an overview of complications after ileostomy or colostomy procedures in children, and to compare outcomes between patients with gastrointestinal motility disorders (i.e. functional constipation, Hirschsprung's disease, pediatric intestinal pseudo-obstruction (PIPO)) and children without motility disorders (including necrotizing enterocolitis, anorectal malformation and inflammatory bowel disease).

Methods: We performed a retrospective study of children who underwent an enterostomy procedure at our institution. The number and type of complications and subsequent reoperations after ostomy formation were determined. Complications were scored using the Clavien–Dindo classification. A complication of \geq grade III-b was considered a high-grade complication.

Results: 129 children with an ileostomy and 61 children with a colostomy were included. Of these, 62 children (32.6%) had motility disorders; functional constipation ($n\!=\!40$), Hirschsprung's disease ($n\!=\!18$) and PIPO ($n\!=\!4$). The total prevalence of complications was 73.2%. Comparing the perioperative data, children with motility disorders significantly more often underwent a laparoscopic procedure (59.7% vs. 10.9%, $p\!=\!0.000$) and had an end stoma-configuration (37.1% vs. 14.1%, $p\!=\!0.000$) as compared to children without motility disorders. Children with motility problems had a higher complication rate (88.7% vs. 65.5%, OR 4.1, 95% CI 1.7–9.8, $p\!=\!0.001$) compared to children without motility problems, and a larger proportion of complications was classified as high-grade complications (61.8% vs. 31.0% $p\!=\!0.002$).

Conclusion: A high complication rate after enterostomy formation was detected. Children with gastrointestinal motility disorders had more and more severe complications as compared to children without motility disorders. Level of evidence: Level III

Type of study: Retrospective comparative study

© 2020 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY license (http://creativecommons.org/licenses/by/4.0/).

Stoma formation is used to manage a wide variety of conditions in children. This includes two types of diverting enterostomies: ileostomies, with the surgically created opening in the small bowel, and colostomies, with the stoma formation in the large bowel. In infants, ostomy formation is often required in children with congenital conditions such as anorectal malformation and Hirschsprung's disease, or acquired conditions such as necrotizing enterocolitis (NEC) [1]. Later in childhood, an ostomy is a treatment option for some children with motility disorders such as intractable functional constipation (FC)

E-mail address: m.h.vriesman@amsterdamumc.nl (M.H. Vriesman).

and pediatric intestinal pseudo obstruction (PIPO), or for patients with inflammatory bowel disease (IBD) [2–6].

After any medical or surgical treatment, complications can occur. Complications after ostomy formation are common and include bleeding, infection, leakage and prolapse of the stoma. In adults, complications have been described to occur in up to 82% of patients [7,8]. In children, several studies in young infants with Hirschsprung's disease, anorectal malformations and NEC describe a stoma-related complication rate between 23% and 42% [9–15]. However, limited data are available in children with other underlying diseases.

In a recent study, a high stoma-related complication rate of 81% was described in a cohort of children with intractable FC [16]. In children with PIPO, complication rates up to 86% have been reported [17]. These studies contributed to the hypothesis that patients with gastrointestinal motility disorders have a higher prevalence of ostomy-related complications compared to patients without motility

 $^{\,\,\}dot{\,}\,\,\,$ Financial support: This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

^{☆☆} Declarations of interest: None.

 $^{^{*}}$ Corresponding author at: Emma Children's Hospital, Amsterdam UMC, University of Amsterdam, H7-250, PO Box 22700, Room C2-312, 1100 DD Amsterdam, the Netherlands. Tel.: +31205665270.

problems. To date, there have been no controlled studies comparing ostomy-related complications in pediatric patients with and without motility disorders.

The aim of this study was to provide an overview of the complication rate after ileostomy or colostomy procedures at our tertiary children's hospital and to compare outcomes between patients with gastrointestinal motility disorders (i.e. FC, Hirschsprung's disease and PIPO) and patients with other indications.

1. Methods

1.1. Study design & population

We performed a retrospective study including all children up to 18 years of age that underwent an ileostomy or colostomy procedure between 2007 and 2017 at a tertiary children's hospital. Children who underwent an appendicocecostomy (Malone), a Chait cecostomy, urostomy or jejunostomy procedure were excluded. In addition, patients who received their primary ostomy at another institution or children for whom perioperative data of the primary surgery were missing were not included in the analysis.

Patients were recruited via an opt-out procedure. All patients received an information letter about the study stating that their anonymized data were to be used for clinical research purposes. Children were excluded from the study if they or their parents declined participation by contacting the research team via mail, email or phone. This protocol was approved by the Medical Ethics Committee of our hospital.

1.2. Surgical procedure

All operations were performed by experienced pediatric surgeons at our institution. Ostomies were classified as either loop, split or end configuration. For all patients, an expert stoma nurse was available for post-surgical stoma care and education.

1.3. Data collection

Relevant patient characteristics were collected from the patient's medical records and obtained from the perioperative notes, follow-up visits and additional emergency room visits. Data were included until their last clinic visit at the (pediatric) gastroenterologist or surgeon, or until the date of death in case patients deceased.

Preoperative data included age, sex, body mass index (BMI), symptoms and diagnosis, treatment(s) prior to surgery and the preoperative diagnostic evaluation results. Perioperative data consisted of information regarding the type of surgery and technical aspects of the ostomy formation. Postoperative data were reviewed to determine if any complications and subsequent reoperations had occurred after the ostomy formation. We defined complications as unexpected and undesired outcomes after surgery, which deviated from the normal postoperative course and were not intrinsic to the procedure. The following complications were evaluated: bleeding, infection, parastomal abscess, peristomal fistula formation, stomal perforation, necrosis (superficial or deep), retraction (>0.5 cm below skin surface), wound dehiscence, parastomal hernia, incisional hernia, fixation problems, prolapse, volvulus, leakage around the ostomy bag, hypergranulation, skin irritation

(dermatitis or excessive erythema), high output (>20 mL/kg/day in infants and >2 L/day in children), stenosis (narrowing during digital examination), troublesome passage of stools (requiring laxative medication), ileus, parastomal or abdominal pain (requiring medication) and respiratory insufficiency.

All complications were scored according to the Clavien–Dindo classification [18]. The Clavien–Dindo classification is a widely used tool to rank postoperative complications and consists of five consecutive grades, ranging from grade I (= no need for pharmacological, surgical, endoscopic and radiological treatment) to grade V (=death) (Table 1). Complications graded as III-b, IV and V were considered to be a high-grade complication.

1.4. Statistical analysis

Data were anonymously stored and analyzed in an SPSS database (IBM SPSS Statistics for Windows, Version 25.0. Armonk, NY: IBM Corp.). Preoperative and postoperative data were compared between patients with severe motility disorders (i.e. FC, Hirschsprung's disease and PIPO) and without motility disorders. We also compared groups that were comparable in terms of age and surgery setting; we therefore compared children with FC versus children with IBD. The study groups were compared by using one-way ANOVA and independent t-test for continuous variables. Analysis of dichotomous variables was performed using Chi-square tests or Fisher's Exact tests. Logistic regression analyses were used to identify risk factors for complications. The Bonferroni correction was applied to account for multiple comparisons. The corresponding p-values accepted for statistical significance are reported.

2. Results

2.1. Study population

A flowchart of the inclusion of patients is depicted in Fig. 1. Out of 255 patients who underwent an ostomy procedure at our institution, 190 children were included in the analyses. Nine parents of patients did not want to participate in the study and chose to opt-out.

Sixty percent of included patients were male. Children underwent ostomy surgery for a variety of indications (Table 2). The majority of patients were less than one year of age at the time of initial surgery and included patients with NEC (15.8%), anorectal malformations (13.7%) and Hirschsprung's disease (9.5%). Indications for older children included FC (21.1%), IBD (8.4%) or trauma (2.6%).

The majority of patients had an ileostomy (n = 129, 67.9%), of which 69 (36.3%) children had a loop, 38 (20%) a split and 22 (11.6%) children an end configuration. A total of 61 patients required a colostomy, of which 25 (13.2%) had a loop, 17 (8.9%) a split and 19 (10%) an end configuration. In all patients with Hirschsprung's disease the stoma was located in normal ganglionic area.

Most children were operated with an open surgical technique (73.2%). Surgery was performed during an emergency setting in 90 children (47.4%) and the ostomy was intended as temporary treatment option in 166 (87.4%) of children. Median hospitalization time after surgery of included patients was 16 days (IQR 9–42). Follow-up ranged from 12 days (patient deceased) to 13 years, and the majority of patients (81.1%) had their primary ostomy closed at the end of follow-

Table 1Complications according to the Clavien–Dindo classification [20].

Grade I	No need for pharmacological,
	surgical, endoscopic and radiological treatment with the exception of drugs such as antiemetics, antipyretics, analgetics, diuretics and physiotherapy
Grade II	Pharmacological treatment other than mentioned in grade I
Grade III-a	Surgical, endoscopic or radiological interventions that do not occur under narcosis
Grade III-b	Surgical, endoscopic or radiological interventions that occur under narcosis
Grade IV	Life-threatening complications that call for IC management
Grade V	Death

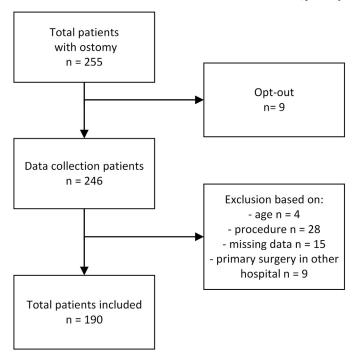


Fig 1. Inclusion of patients.

up. Twenty-one patients (11.1%) required a second ostomy operation. Four patients died (2.1%); none of these deaths were directly related to ostomy formation or closure.

2.2. Complication rate

In total, 139 out of 190 children (73.2%) had at least one complication after their ostomy formation. Patients experienced a median number of 2 complications (IQR 1–4). According to the Clavien–Dindo classification, the highest grade of complication was grade I in 66 patients (34.7%), grade II in 11 (5.8%), grade III-a in 2 (1.1%), grade III-b in 59 (31.1%) and grade IV in 1 (0.5%) patient. The most common complications were leakage (28.4%), abdominal pain (26.8%), and high output (21.6%, Table 3). Ostomy-related complications led to 126 surgical

Table 2 Indication ostomy (n = 190).

	Total group ($n = 190$)
Functional constipation	40 (21.1%)
Necrotizing enterocolitis	30 (15.8%)
Anorectal malformation	26 (13.7%)
Hirschsprung's disease	18 (9.5%)
Inflammatory bowel disease	16 (8.4%)
Surgical complication after previous abdominal surgery§	13 (6.8%)
Small bowel atresia	12 (6.3%)
Cystic fibrosis	6 (3.2%)
Trauma	5 (2.6%)
PIPO	4 (2.1%)
Volvulus	1 (0.5%)
Other¥	19 (10.0%)

PIPO = pediatric intestinal pseudo-obstruction

Table 3 Overview of all complications.

Complication	Total group ($n=190$), n (%)
Leakage	54 (28.4%)
Abdominal pain	51 (26.8%)
High output	41 (21.6%)
Skin irritation	39 (20.5%)
Troublesome passage stools requiring laxatives	35 (18.4%)
Prolapse	31 (16.3%)
Bleeding	27 (14.2%)
Ileus	24 (12.6%)
Infection	23 (12.1%)
Wound dehiscence	19 (10.0%)
Retraction	14 (7.4%)
Stenosis	12 (6.3%)
Stomal perforation	10 (5.3%)
Peristomal fistula formation	8 (4.2%)
Parastomal hernia	8 (4.2%)
Volvulus	7 (3.7%)
Parastomal abscess	5 (2.6%)
Deep necrosis	5 (2.6%)
Fixation problems	5 (2.6%)
Hypergranulation	5 (2.6%)
Superficial necrosis	4 (2.1%)
Incisional hernia	2 (1.1%)
Respiratory insufficiency	1 (0.5%)

revisions; the most common indications for surgical revisions were ileus (n=15), abdominal pain (n=15) and prolapse (n=13).

Comparing the demographical data of patients with and without complications, no differences were found with respect to age, sex and BMI of children. For this analysis, after Bonferroni adjustment, a p<0.006 was considered statistically significant. The type of ostomy (i.e. ileostomy or colostomy), surgical setting (i.e. elective or emergency) and duration of ostomy were similar between both groups. Comparing stoma configurations, children with a complication significantly less often had a split stoma configuration (47.8% vs. 22.9%, 95% CI 0.2–0.7, p=0.001); no differences were found with respect to end and loop configurations. Significantly more patients with a complication underwent a laparoscopic procedure (31.9% vs. 10.9%, OR 3.6, 95% CI 1.4–9.0, p=0.005) as compared to patients without complications.

2.3. Complication rate of patients with motility disorders versus other

The number and type of complications were compared between patients with and without motility disorders. A total of 62 children had preoperative evidence of gastrointestinal motility problems, including FC (n=40), Hirschsprung's disease (n=18) and PIPO (n=4) (Table 4). Comparing the stoma configuration, significantly more end configurations were found in patients with motility disorders (37.1% vs. 14.1%, p=0.000), significantly fewer children with motility disorders had a split configuration (12.9% vs. 36.7%, p=0.001) and the number of patients with a loop configuration was similar between patients with and without motility problems (50.0% vs. 49.2%, p=0.920).

Comparing the complication rate, significantly more patients with a motility disorder had at least one complication (88.7% vs. 65.5%, OR 4.1, 95% CI 1.7–9.8, p=0.001) (Table 5). Moreover, patients with motility disorders more frequently experienced a high-grade complication (i.e. grade III-b or higher) compared to patients without motility disorders (61.8% vs. 31%, p=0.002) and had a longer median duration of hospital stay after surgery (24 vs. 11 days, p=0.000). Adjusted for follow-up duration and receiving multiple stomas, having a motility disorder remained an independent risk factor for complications after ostomy formation (OR 4.3, 95% CI 1.7–10.7, p=0.001).

In children with motility disorders, the most prevalent complications included abdominal pain (56.5%), troublesome passage of stools (37.1%), leakage (35.5%) and ileus (27.4%). In children without motility disorders, the most prevalent complications included leakage (25%),

we microcolon (n = 3), meconium ileus of unknown origin (n = 2), familial adenomatous polyposis (n = 2), perforated appendicitis (n = 1), rectovaginal fistula (n = 1), spondylodiscitis with rectosigmoid fistula (n = 1), E. coli sepsis with perforation bowel (n = 1), solitary rectal ulcer (n = 1), rhabdomyosarcoma with rectal obstruction (n = 1), caudal regression syndrome (n = 1), Meckel diverticulum (n = 1), rectal prolapse (n = 1), bowel obstruction of unknown origin (n = 1), GUCY2C mutation (n = 1), blow-out sigmoid due to stenosis of unknown origin (n = 1).

 $^{^{\}S}=$ anastomotic leakage (n = 5), bowel perforation (n = 3), intestinal ischemia (n = 3), intestinal obstruction (n = 2).

Table 4 Comparison of different motility disorders.

	Functional constipation $(n=40)$	Hirschsprung's disease ($n=18$)	PIPO (n=4)		
Age, median (IQR)	11.5 years	13 days	10.8 years		
	(6.2-16.8 years)	(3 days-2.4 years)	(2.6–15.5 years)		
Males, n (%)	12 (30.0%)	15 (83.3%)	4 (100%)		
Perioperative data					
Ileostomy, n (%)	29 (72.5%)	12 (66.7%)	4 (100%)		
Emergency setting <i>n</i> (%)	4 (10.0%)	10 (55.6%)	1 (25.0%)		
Laparoscopic procedure, n (%)	33 (82.5%)	3 (16.7%)	1 (25.0%)		
Loop configuration, n (%)	22 (55.0%)	8 (44.4%)	1 (25.0%)		
Split configuration, n (%)	3 (7.5%)	4 (22.2%)	1 (25.0%)		
End configuration, n (%)	15 (37.5%)	6 (33.3%)	2 (50.0%)		
Temporary intention, n (%)	27 (67.5%)	16 (88.9%)	1 (25.0%)		
	Postoperative data	1			
Days of hospital stay, median (IQR)	9 (6-16)	15 (9–19)	45 (6-45)		
Ostomy closure, n (%)	21 (52.5%)	17 (94.4%)	0 (-)		
Multiple ostomies, n (%)	8 (20.0%)	4 (22.2%)	1 (25.0%)		
Complication, n (%)	38 (95.0%)	13 (72.2%)	4 (100%)		
High-grade complication, n (%)	27 (71.1%)	3 (16.7%)	4 (100%)		
Reoperations, median (IQR)	2 (0-3)	1 (1–2)	8 (2-10)		
Readmissions, median (IQR)	3 (0-5)	1 (1–2)	11 (4–13)		

IQR = interquartile range, PIPO= pediatric intestinal pseudo-obstruction

high output of the stoma (21.1%), and skin irritation (18.8%). Comparing the type of complication, significantly more patients with motility disorders had troublesome passage of stools, abdominal pain and ileus (all p<0.002, Table 6).

When discarding the complications abdominal pain and troublesome passage of stools, both common symptoms in patients with FC and PIPO, the number of patients with complications remained higher in the children with motility problems as compared to patients without motility problems (75.8% vs. 62.5%).

We compared two groups of children with similar age and surgery setting characteristics: patients with FC ($n\!=\!40$) and children with IBD ($n\!=\!16$). In both groups, ileostomy was the most common stoma type (72.5% of FC patients vs. 87.5% of IBD patients, NS). Significantly more patients with FC had one or more complications compared to patients with IBD (95.0% vs. 62.5%, $p\!=\!0.005$) and more patients with FC had a high-grade complication as compared to the children with IBD (71.1% vs. 10%, $p\!=\!0.000$).

3. Discussion

In this study, we described the prevalence and severity of complications after ostomy formation in a large pediatric cohort using a predefined standardized complication-grading tool; we also compared complication rates between pediatric patients with and without motility disorders. In our heterogeneous patient cohort, including children with functional and organic gastrointestinal disorders, the total complication rate after ileostomy and colostomy procedures was 73.2% and one third of patients had a high-grade complication leading to surgical revision of the ostomy. Fecal leakage was the most frequently encountered complication.

Although the ostomy-related complication rate that we found is substantial, it is comparable to results found in adult literature. Previous prospective studies in adults have described an ostomy-related complication rate up to 82% [7,8]. Reported risk factors for complications in adults included a high BMI [19] and emergency surgery [19]; in our

Table 5Comparison of patients with and without motility disorders.

	Motility disorders (n=62)	No motility disorders (n=128)	p-value
Age, median (IQR)	8.1 years	18 days	0.000*
	(1.4–15.5 years)	(2 days -4.1 years)	
Males, n (%)	31 (50.0%)	83 (64.8%)	0.050
	Perioperative data		
Ileostomy, n (%)	45 (72.6%)	84 (65.6%)	0.336
Emergency setting, n (%)	15 (24.2%)	75 (58.6%)	0.000^*
Laparoscopic procedure n (%)	37 (59.7%)	14 (10.9%)	0.000^{*}
Loop configuration, $n(%)$	31 (50.0%)	63 (49.2%)	0.000^{*}
Split configuration, n (%)	8 (12.9%)	47 (36.7%)	
End configuration, n (%)	23 (37.1%)	18 (14.1%)	
Temporary intention, n (%)	44 (71.0%)	112 (95.3%)	0.000^*
	Postoperative data		
Days of hospital stay, median (IQR)	11 (7–18)	24 (11-49)	0.000*
Ostomy closure, n (%)	38 (61.3%)	116 (90.6%)	0.000^*
Multiple ostomies, n (%)	13 (21.0%)	8 (6.3%)	0.002*
Complication, $n(%)$	55 (88.7%)	84 (65.5%)	0.001*
High-grade complication, n (%)	34/55 (61.8%)	26/84 (31.0%)	0.002*
Reoperations, median (IQR)	2 (1–3)	1 (1-1)	0.000^*
Readmissions, median (IQR)	2 (1–5)	1 (1–1)	0.000^*
Follow-up in months, median (IQR)	35 (11–83)	18 (5–42)	0.001*

⁼ statistically significant corrected by using the Bonferroni correction for 15 comparisons (p<0.003).

Table 6Comparison of type of complication between patients with and without motility disorders.

	Motility disorders $(n=62)$	No motility disorders ($n = 128$)	p-value
Leakage	22 (35.5%)	32 (25.0%)	0.133
Abdominal pain	35 (56.5%)	16 (12.5%)	0.000*
High output	14 (22.6%)	27 (21.1%)	0.815
Skin irritation	15 (24.2%)	24 (18.8%)	0.444
Troublesome passage stools requiring laxatives	23 (37.1%)	12 (9.4%)	0.000*
Prolapse	12 (19.4%)	19 (14.8%)	0.430
Bleeding	9 (14.5%)	18 (14.1%)	1.000
Ileus	17 (27.4%)	7 (5.5%)	0.000^{*}
Infection	10 (16.1%)	13 (10.2%)	0.237
Wound dehiscence	4 (6.5%)	15 (11.7%)	0.257
Retraction	8 (12.9%)	6 (4.7%)	0.071
Stenosis	7 (11.3%)	5 (3.9%)	0.061
Stomal perforation	5 (8.1%)	5 (3.9%)	0.299
Peristomal fistula formation	3 (4.8%)	5 (3.9%)	0.717
Parastomal hernia	4 (6.5%)	4 (3.1%)	0.441
Volvulus	6 (9.7%)	1 (0.8%)	0.005
Parastomal abscess	2 (3.2%)	3 (2.3%)	0.662
Deep necrosis	4 (6.5%)	1 (0.8%)	0.040
Fixation problems	2 (3.2%)	3 (2.3%)	0.662
Hypergranulation	3 (4.8%)	2 (1.6%)	0.332
Superficial necrosis	1 (1.6%)	3 (2.3%)	1.000
Incisional hernia	1 (1.6%)	1 (0.8%)	0.547
Respiratory insufficiency	0 (-)	1 (0.8%)	1.000

 $^{^{*}=}$ statistically significant corrected by using the Bonferroni correction for 23 comparisons (p<0.002).

study we did not find significant differences related to these factors. The indication for ostomy formation, however, differs substantially between adults and children; in the adult literature, the majority of patients were elderly patients with malignancies, with high rates of comorbidity and medication use, whereas our study included mostly young infants with congenital anomalies. Consequently, it is difficult to compare studies and the type of complications between children and adults.

In comparison to the available pediatric literature [9–15], we found a higher prevalence of ostomy-related complications. Only one other study found a similar rate of complications (80.5%) after colostomy surgery in young infants with anorectal malformations, Hirschsprung's disease and trauma [20]. The authors explained this high complication rate by the nonavailability of an expert stoma nurse and poor socioeconomic level of included families. However, these factors did not apply to our cohort.

A possible explanation for the high complication rate found in our cohort could be our definition of complications. We defined complications as unexpected and undesired outcomes occurring after surgery, not intrinsic to the surgical procedure itself. The definition of complications and the Clavien-Dindo classification system have had an important effect on the definitions reported and the severity attributed to them. The use of different definitions and classification systems may explain some of the differences between our results and previously published reports. We systematically reviewed charts to search for a large number of predefined complications. While most previously published studies only reported "classical" complications related to the ostomy itself, such as prolapse or bleeding, we also included gastrointestinal symptoms such as abdominal pain and troublesome passage of stools. Although not life-threatening, these negative outcomes after surgery often required medical treatment or further surgery. As an example, in our cohort, abdominal pain was one of the most common indications for surgical revision of the stoma. These complications accounted for a number of healthcare visits and are likely to have significantly affected the quality of life of included children.

Another possible explanation for the high complication rate found in our study is the large number of patients with motility disorders included in our cohort (32.6%). We hypothesized that patients with motility disorders more frequently experienced complications after ostomy

procedures than children without underlying motility disorders. Not only did we find a higher complication rate in children with motility disorders, we also found a higher prevalence of severe complications as compared to children without motility disorders.

Several limitations should be taken into account when interpreting these results. Patients with motility disorders had a significantly longer follow-up and often required a second ostomy, possibly contributing to a higher risk of complications. However, adjusted for these factors, motility disorders remained to be significantly associated with the occurrence of more complications.

In addition, in our cohort, significant differences in perioperative characteristics between patients with and without motility disorders were reported, making it challenging to compare both groups and draw firm conclusions. Significantly more children with motility disorders underwent a laparoscopic procedure in comparison with children without motility disorders. Moreover, significantly more children with motility disorders had end stoma-configuration and fewer children had a split configuration as compared to children without motility disorders. In line with our results, split stoma configuration is reported to be associated with a lower complication rate in several studies [9,12,13]. It could therefore be hypothesized that the high complication rate in children with motility disorders was explained by the type of procedure rather than their underlying diagnosis.

On the other hand, the high complication rate in children with motility disorders could have been related to the disease itself rather than a consequence of the surgery. When comparing the children with and without motility disorders, the prevalence of "classical" stoma-related complications was not significantly different between the two groups. Analyzing the data in more detail, the higher complication rate in the motility group was mostly attributed to the higher prevalence of gastrointestinal complications (i.e., abdominal pain, troublesome passage of stools, ileus, volvulus). Although ileostomy and colostomy are effective surgical strategies for patients with intractable FC. Hirschsprung's disease and PIPO, surgery does not cure the underlying motility problems throughout the remaining gastrointestinal tract. In fact, in patients with FC, Hirschsprung's disease and PIPO, evidence of upper gut motility problems has been reported [21-24]. Patients with Hirschsprung's disease, although diverted into normal ganglionic colon after their stoma formation, were therefore analyzed as part of the motility disorders group. However, it is not surprising that motility related complications, such as ileus, high output stoma, abdominal pain and troublesome passage of stools, were prevalent in these patients. Previous studies in patients with PIPO reported high output and prolonged paralytic ileus as common postsurgical problems after ostomy formation [3,25,26]. Also, in patients with functional gastrointestinal disorders including FC, low pain threshold and visceral hypersensitivity have been described owing to dysregulation of the brain-gut axis [27,28], potentially contributing to the higher rates of postoperative abdominal pain in children with motility disorders. Moreover, preexisting symptoms of abdominal pain have been reported as an important risk factor for chronic abdominal pain after abdominal surgery [29]. However, to account for this, we also calculated the complication rate without abdominal pain and troublesome passage of stools. The complication rate of the total population remained high, with the persistence of higher complication rates in patients with motility disorders.

Several other limitations should be noted. Missing data and the exclusion of patients possibly led to the underreporting of complications. Moreover, our cohort consisted of a heterogeneous group of patients with a wide variety of age and indications for ostomy formation, making it challenging to draw firm conclusions about our entire cohort. We therefore grouped together patients with similar age and indication in our analysis to compare the prevalence of complications. Consequently, patient groups were relatively small. For example, owing to the limited number of patients with PIPO in our cohort, previously reported factors such as high rates of stoma prolapse in these children could not be confirmed [17,30,31]. Surgical errors should also be taken into account. For

example, the high number of ileus as an indication for surgical revisions could be procedure-related. The opening in the posterior fascia may have been too tight or small, leading to stenosis. Moreover, owing to the retrospective design of our study, other previously reported risk factors for complications such as parental factors could not be determined [20,32]. In addition, nutritional status and type of nutrition (oral, tube or parenteral) of included children changed frequently over the follow-up period and could not be identified as risk factors for complications.

In conclusion, in this heterogeneous cohort of children with ostomy formation, we found a remarkably high ostomy-related complication rate. Patients with gastrointestinal motility disorders had a higher complication rate and more severe complications in comparison to the children without gastrointestinal motility disorders. Our results confirm that surgical strategies in patients with motility disorders should be seen a treatment of last resort and high rates of complications should be expected and taken into consideration when choosing the type of surgical management. However, owing to the retrospective design of the study and significant differences between patients with and without motility disorders with respect to type and configuration of the stoma, our results should be interpreted with care. Future prospective studies including homogenous patient cohorts of children with and without motility disorders, standardized follow-up visits and registration of complications after surgery are warranted to confirm our results.

References

- Paediatric stoma care global best practice guidelines for neonates, children and teenagers, https://www.crohn-colitis.nl/wp-content/uploads/2019/03/Internationalerichtlijn-voor-kinderstomazorg.pdf. [Accessed 30 July 2019].
- [2] Siminas S, Losty PD. Current surgical management of pediatric idiopathic constipation. Ann Surg. 2015;262(6):925–33. https://doi.org/10.1097/SIA.000000000001191.
- [3] Thapar N, Saliakellis E, Benninga MA, et al. Paediatric intestinal pseudo-obstruction: evidence and consensus-based recommendations from an ESPGHAN-led expert group. J Pediatr Gastroenterol Nutr. 2018;66(6):991–1019. https://doi.org/10. 1097/MPG.0000000000001982.
- [4] Amii-Dias J, Kolacek S, Turner D, et al. Surgical management of crohn disease in children. J Pediatr Gastroenterol Nutr. 2017;64(5):818–35. https://doi.org/10.1097/MPG.000000000001562.
- [5] Turner D, Ruemmele FM, Orlanski-Meyer E, et al. Management of paediatric ulcerative colitis, part 1: ambulatory care-an evidence-based guideline from European Crohn's and Colitis Organization and European Society of Paediatric Gastroenterology, Hepatology and Nutrition. J Pediatr Gastroenterol Nutr. 2018;67(2):257–91. https://doi.org/10.1097/MPG.000000000002035.
- [6] Tamura R, Jaffray B. Outcomes of colonic resection for chronic idiopathic constipation in childhood. J Pediatr Surg. November 2019. https://doi.org/10.1016/j.jpedsurg.2019.10.047.
- [7] Formijne Jonkers HA, Draaisma WA, Roskott AM, et al. Early complications after stoma formation: a prospective cohort study in 100 patients with 1-year followup. Int J Color Dis. 2012;27(8):1095–9. https://doi.org/10.1007/s00384-012-1413-y.
- [8] Malik T, Lee MJ, Harikrishnan AB. The incidence of stoma related morbidity a systematic review of randomised controlled trials. Ann R Coll Surg Engl. 2018;100(7): 501–8. https://doi.org/10.1308/rcsann.2018.0126.
- [9] Youssef F, Arbash G, Puligandla PS, et al. Loop versus divided colostomy for the management of anorectal malformations: a systematic review and meta-analysis. J Pediatr Surg. 2017;52(5):783–90. https://doi.org/10.1016/j.jpedsurg.2017.01.044.
- [10] Steinau G, Ruhl KM, Hörnchen H, et al. Enterostomy complications in infancy and childhood. Langenbeck's Arch Surg. 2001;386(5):346–9. https://doi.org/10.1007/ s004230100243.

- [11] Bethell G, Kenny S, Corbett H. Enterostomy-related complications and growth following reversal in infants. Arch Dis Child Fetal Neonatal Ed. 2017;102(3):F230–4. https://doi.org/10.1136/archdischild-2016-311126.
- [12] van den Hondel D, Sloots C, Meeussen C, et al. To split or not to split: colostomy complications for anorectal malformations or Hirschsprung disease: a single center experience and a systematic review of the literature. Eur J Pediatr Surg. 2013;24 (01):061–9. https://doi.org/10.1055/s-0033-1351663.
- [13] Oda O, Davies D, Colapinto K, et al. Loop versus divided colostomy for the management of anorectal malformations. J Pediatr Surg. 2014;49(1):87–90. https://doi.org/ 10.1016/j.jpedsurg.2013.09.032.
- [14] Aguayo P, Fraser JD, Sharp S. St. Peter SD, Ostlie DJ. Stomal complications in the newborn with necrotizing enterocolitis. J Surg Res. 2009;157(2):275–8. https://doi.org/ 10.1016/i.iss.2009.06.005.
- [15] Pena A, Migotto-Krieger M, Levitt MA. Colostomy in anorectal malformations: a procedure with serious but preventable complications. J Pediatr Surg. 2006;41(4): 748–56. https://doi.org/10.1016/j.jpedsurg.2005.12.021.
- [16] Kuizenga-Wessel S, Koppen IJN, Zwager LW, et al. Surgical management of children with intractable functional constipation; experience of a single tertiary children's hospital. Neurogastroenterol Motil. 2017;29(5):e13005. https://doi.org/10.1111/nmo.13005.
- [17] Kim H-Y, Kim J-H, Jung S-E, et al. Surgical treatment and prognosis of chronic intestinal pseudo-obstruction in children. J Pediatr Surg. 2005;40(11):1753–9. https://doi.org/10.1016/j.jpedsurg.2005.07.046.
- [18] Dindo D, Demartines N, Clavien P-A. Classification of surgical complications: a new proposal with evaluation in a cohort of 6336 patients and results of a survey. Ann Surg. 2004;240(2):205–13. https://doi.org/10.1097/01.SLA.0000133083.54934.AE.
- [19] Arumugam PJ, Bevan L, Macdonald L, et al. A prospective audit of stomas—analysis of risk factors and complications and their management. Color Dis. 2003;5 (1):49–52
- [20] Çiğdem MK, Onen A, Duran H, et al. The mechanical complications of colostomy in infants and children: analysis of 473 cases of a single center. Pediatr Surg Int. 2006;22(8):671–6. https://doi.org/10.1007/s00383-006-1718-4.
- [21] Di Nardo G, Di Lorenzo C, Lauro A, et al. Chronic intestinal pseudo-obstruction in children and adults: diagnosis and therapeutic options. Neurogastroenterol Motil. 2017;29(1):e12945. https://doi.org/10.1111/nmo.12945.
- [22] Tomita R, İkeda T, Fujisaki S, et al. Upper gut motility of Hirschsprung's disease and its allied disorders in adults. Hepato-Gastroenterology. 2003;50(54):1959–62.
- [23] Boccia G, Buonavolontà R, Coccorullo P, et al. Dyspeptic symptoms in children: the result of a constipation-induced cologastric brake? Clin Gastroenterol Hepatol. 2008;6(5):556–60. https://doi.org/10.1016/j.cgh.2008.01.001.
- [24] Tjeerdsma HC, Smout AJPM, Akkermans LMA. Voluntary suppression of defecation delays gastric emptying. Dig Dis Sci. 1993;38(5):832–6. https://doi.org/10.1007/ BF01295908
- [25] Heneyke S, Smith VV, Spitz L, et al. Chronic intestinal pseudo-obstruction: treatment and long term follow up of 44 patients. Arch Dis Child. 1999;81(1):21–7. https://doi. org/10.1136/adc.81.1.21.
- [26] Rolston DD, Hunt JB, Fairclough PD, et al. Jejunal water and sodium secretion occurs in chronic idiopathic intestinal pseudo-obstruction. J Clin Gastroenterol. 1990;12(2): 153–6. https://doi.org/10.1097/00004836-199004000-00008.
- [27] Azpiroz F. Hypersensitivity in functional gastrointestinal disorders. Gut. 2002;51 (Supplement 1):i25–8. https://doi.org/10.1136/gut.51.suppl_1.i25.
- [28] Drossman DA, Hasler WL. Rome IV-functional GI disorders: disorders of gut-brain interaction. Gastroenterology. 2016;150(6):1257–61. https://doi.org/10.1053/j. gastro.2016.03.035.
- [29] Strik C, van den Beukel B, van Rijckevorsel D, et al. Risk of pain and gastrointestinal complaints at 6 months after elective abdominal surgery. J Pain. 2019;20(1):38–46. https://doi.org/10.1016/j.jpain.2018.07.010.
- [30] Irtan S, Bellaïche M, Brasher C, et al. Stomal prolapse in children with chronic intestinal pseudoobstruction: a frequent complication? J Pediatr Surg. 2010;45(11): 2234–7. https://doi.org/10.1016/j.jpedsurg.2010.06.022.
- [31] Sparks EA, Velazco CS, Fullerton BS, et al. Ileostomy prolapse in children with intestinal dysmotility. Gastroenterol Res Pract. 2017;2017;7182429. https://doi.org/10.1155/2017/7182429.
- [32] Persson E, Berndtsson I, Carlsson E, et al. Stoma-related complications and stoma size — a 2-year follow up. Color Dis. 2010;12(10):971–6. https://doi.org/10.1111/j. 1463-1318.2009.01941.x.